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Hand Assessment for Infants

-Development, internal scale validity,
reliability and normative reference values

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Hand Assessment for Infants -Development, internal scale validity, reliability and normative reference values

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To my wonderful children Hedda and Gry

“Behind every statistic there’s a human beating heart”

- Stephen Fry

ABSTRACT

Infants use their hands to explore the world and neural event in the developing brain can affect the infant's ability to use the hands. An infant that has suffered a brain lesion may show clinical signs of unilateral cerebral palsy as early as at three months of age, however, to quantify an asymmetric hand use and evaluate the results of early interventions there is a need for assessment tools producing valid and reliable measures.

The aims of the Studies in this thesis were to construct an assessment tool measuring bimanual goal oriented hand use in infants 3-12 months of age, to investigate its psychometric properties for infants with clinical signs of unilateral cerebral palsy as well as to create normative reference values.

Study I describes the development of the Hand Assessment for Infants (HAI). The HAI is an assessment tool that constitutes of 17 observable items all measuring different aspects of goal oriented hand use. Twelve of the items are unimanual, scored for the right and the left hand separately, and five items are bimanual where one score is given. All items are scored on a three-point rating scale (0-2) and the scores of the unimanual items are summed to the Each Hand Sum score (EaHS) with a range of 0-24 raw scores. The EaHS of the better functioning hand and the lesser functioning hand is used to calculate an asymmetry index where a higher percentage indicates a larger asymmetry. The results of all the items are added to the Both Hands Sum score (0-58 raw scores) that has been converted to logits through the Rasch measurement model analysis to become the Both Hands Measure (BoHM), an interval scale of 0-100 HAI-units and the main outcome of the HAI.

The internal construct validity of the HAI was examined by the use of Rasch measurement model analysis and we found that the HAI has a unidimensional construct and good fit to the Rasch model requirements for both items and persons. A high separation index shows that the HAI can separate between infants of different levels of ability.

In Study II normative reference values were created through compiling 489 HAI assessments of typically developing infants 3-10 months. Results show that typically developing infants improve their bimanual hand use, as measured by the HAI, at a rapid pace and that a clear majority of typically developing infants does not demonstrate an asymmetrical hand use. We can now compare the results of an infant with suspected signs of unilateral CP to the reference values to identify infants with atypical hand use.

In Study III the reliability of the measures of the HAI was investigated both for test-retest and inter rater reliability. Both test-retest and interrater reliability of the outcomes of the HAI was excellent as shown by the range of the reliability coefficient (Intraclass Correlation Coefficient) 0.97-0.99. For all individual items reliability was good or excellent with an ICC range of 0.81-0.99. The smallest detectable difference of the HAI is 2.36 meaning that a difference in results in 3 HAI-units can be considered a true change.

In summary the studies in this thesis shows that the HAI provides valid and reliable measures of the object related hand use of infants with signs of unilateral Cerebral Palsy. The HAI can be used to identify a possible asymmetry and differentiate infants with an atypical hand use as well as to quantify asymmetric hand use over time in infants with clinical signs of unilateral cerebral palsy.

Key words: Unilateral CP, hand use, infants, assessment, psychometric properties

LIST OF SCIENTIFIC PAPERS

This thesis is based on the following three paper, which will be referred to by their roman numerals:

- I. Development of the Hand Assessment for Infants: evidence of internal scale validity. Krumlinde-Sundholm L, Ek L, Sicola E, Sjöstrand L, Guzzetta A, Sgandurra G, Cioni G, Eliasson AC. Dev Med Child Neurol. 2017 Oct 6.
- II. Hand Assessment for Infants: normative reference values. Ek L, Eliasson AC, Sicola E, Sjöstrand L, Guzzetta A, Sgandurra G, Cioni G, Krumlinde-Sundholm L. Dev Med Child Neurol. 2019 Feb 4
- III. Interrater and test-retest reliability of the Hand Assessment for Infants, HAI. Ek L, Ullenhag A, Eliasson AC, Krumlinde-Sundholm L. In manuscript

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LIST OF ABBREVIATIONS

AHA	Assisting Hand Assessment
BoHM	Both Hands Measure
BoHS	Both Hands Sum score
BSITD	Bayley Scale of Infant and Toddler Development
CP	Cerebral Palsy
CTT	Classical Test Theory
DAYC	Developmental Assessment of Young Children
DIF	Differential Item Functioning
EaHS	Each Hand Sum score
eTIPS	Early Therapy in Perinatal Stroke
GRAB	Grasp and Reach Assessment of Brisbane
HAI	Hand Assessment for Infants
ICC	Intraclass Correlation Coefficient
IMP	Infant Motor Profile
Infant	A child aged 0-12 months of age
IRT	Item Response Theory
MAI	Movement Assessment of Infants
Mini-AHA	Mini Assisting Hand Assessment
MRI	Magnetic Resonance Imaging
PCA	Principal Component Analysis
PDMS	Peabody Developmental Motor Scales
RCT	Randomized Controlled Trial
SD	Standard Deviation
SDD	Smallest Detectable Difference
SEM	Standard Error of Measurement
SOMP	Structured Observation of Motor Performance
Standards	Standards for educational and psychological testing
TIME	Toddler and Infant Motor Evaluation
WMI	White Matter Injury

1 INTRODUCTION

1.1 GENERAL INTRODUCTION

Cerebral palsy (CP) is the most common cause of physical disability for children (Rosenbaum et al., 2007). Children with unilateral CP have one affected hand that is poorer functioning that affects the child's bimanual performance (Charles, 2008). These difficulties in bimanual performance influences the abilities to carry out activities of daily living (Skold, Josephsson, & Eliasson, 2004) as well as the possibility to become independent in self-care throughout the whole life (Russo, Skuza, Sandelance, & Flett, 2019).

There is a wide consensus of the importance of setting the diagnosis of CP early during infancy, both to provide cerebral palsy specific interventions to take advantage of the rapid neurodevelopment during infancy and to provide the appropriate support for the families (Basu, Pearse, Kelly, Wisher, & Kisler, 2014; Novak et al., 2017). There is also knowledge that asymmetric hand use, that can be seen as early as at three months of age, is one of the first signs of unilateral CP (Guzzetta et al., 2010).

Despite this, until recently there has not been an assessment tool that could quantify a possible asymmetry in hand use which is an early sign of unilateral CP. Nor has there been an assessment tool to evaluate the effects of early motor interventions (Krumlinde-Sundholm, Ek, & Eliasson, 2015). The focus of this thesis will be the development of such an instrument, the Hand Assessment for infants (HAI) as well as the evaluation of its psychometric properties.

1.2 TYPICAL DEVELOPMENT OF HAND USE

The typically developing infant shows a rapid development of hand use during the first year. This development is driven by the infants internal will to explore the world and is influenced by the opportunities they are given to use their hands in this challenge (Henderson & Pehoski, 2006; von Hofsten, 2004).

1.2.1 Reaching

Already at birth infants use goal oriented hand and arm movements to reach for objects that they see (van der Meer, van der Weel, & Lee, 1995; von Hofsten, 1982) and as they start to gain more control of their arm and hand movements the amount of goal directed movements increase (von Hofsten, 1984). At four months of age typically developing infants are consistently reaching for objects to grasp them (Berthier, Clifton, McCall, & Robin, 1999).

Initially the infant most often reaches bimanually towards all objects placed in midline (Pehoski, 2006; Sgandurra et al., 2012) but starting at 4 months of age infants begin to discriminate between objects based on the size to reach unimanually for smaller objects and bimanually for larger objects (Sgandurra et al., 2012; Van Hof, Van der Kamp, & Savelsbergh, 2002). At 5 months of age typically developing infants consistently adjust their reach to the size of the object (Van Hof et al., 2002).

Typically developing infants do not cross midline to reach for objects before six months of age and after that point can cross midline during both bimanual and unimanual reach for toys (Van Hof et al., 2002). However, typically developing infants usually reach for objects with the hand that is closest (Morange & Bloch, 1996).

1.2.2 Grasping and holding

At birth infants have a grasp reflex and can hold on to objects like a finger but the reflex decreases during the first few months as the infant starts to gain control of their motor actions. At about four months of age the infant develops control of voluntarily grasp an object that is presented close to the hand (Charles, 2008; Gesell, 1934; Gesell, Thompson, Amatruda, & Yale University. Clinic of Child Development., 1934) and at four months of age the infant holds objects in midline with both hands (Case-Smith, 1995; Pierce, Munier, & Myers, 2009).

At five months of age infants start to adjust their grasp to the size of the object (Newell, McDonald, & Baillargeon, 1993; von Hofsten & Ronqvist, 1988) and they also start to adjust the position of the hand to fit the position of the object that they intend to grasp (Lockman, Ashmead, & Bushnell, 1984; Witherington, 2005). At five months of age infants primarily adjust the arm and hand through tactile feedback after the hand gets in to contact with the object (Lockman et al., 1984). During the next two months there is a rapid development as the infant increasingly adjust the grasp based on visual feedback, having already adjusted the hand and arm position when touching it (Witherington, 2005). This skill is refined further through the first year of life as the infant improves the ability to adjust the grasp to the objects (Pehoski, 2006).

The first grasp that the infant uses is a palmar grip where objects are held towards the palm using all four fingers (Pehoski, 2006). As the infant's dexterity develops the grasp becomes more distal and at nine or ten months of age the infant can pick up small objects using only the thumb and finger (pincher grasp) (Halverson, 1931; Sgandurra et al., 2012).

1.2.3 Hand preference

The development of hand preference can follow different trajectories but some infants have a stable handedness from very early age (Nelson, Campbell, & Michel, 2013). Hand preference emerges earlier if the infant has a right hand preference (6 months) than a left hand preference (8 months) and a right hand preference seems to be more stable over time (Michel, Campbell, Marcinowski, Nelson, & Babik, 2016). Hand preference is first demonstrated by the infant reaching for toys more frequently with one hand (Ferre, Babik, & Michel, 2010). Hand preference in unimanual manipulation is demonstrated at about 10-11 months of age (J. M. Campbell, Marcinowski, Babik, & Michel, 2015) and is manifest in role differentiated hand use at about 18 months of age (Nelson et al., 2013).

1.2.4 Manipulating objects

At about six or seven months of age the typically developing infant starts to consciously release objects to the other hand or to a surface and at this age the infant can coordinate the hands to start to move toys between the hands (Gesell et al., 1934; Pehoski, 2006; Pierce et al., 2009).

Before the age of about 10-12 months the infant primarily uses the hands symmetrically in bimanual hand use for example to transfer a toy or bang them together (Pehoski, 2006) even though aspects of role differentiated hand use is seen as early as seven months of age (Charles, 2008; Kimmerle, Ferre, Kotwica, & Michel, 2010). However, the skill of using role differentiated hand use to explore objects, meaning that the hands do different things is not acquired during an infant's first year of life (Kimmerle et al., 2010).

1.2.5 Factors that influence development of fine motor skills

The development of hand use does not occur in isolation but rather in complex interaction with the senses, for example vision, touch and proprioception, as well as postural control and cognition (Case-Smith, 1995; Charles, 2008).

Vision is central for the infant's awareness of the hands as well as for the process of learning to use them (Hyvarinen, Walther, Jacob, Chaplin, & Leonhardt, 2014) and an infant's ability to successfully reach for and grasp an object is controlled by vision to a large extent (von Hofsten & Ronnqvist, 1988).

The infant's postural control influences the development of reaching for objects as well as manipulating them (Charles, 2008). As the infants' ability to sit independently develops so does

the amount and quality of reach for items (Rachwani, Santamaria, Saavedra, & Woollacott, 2015).

The infant's cognitive development influences all aspects of the development of object related hand use (Exner & Henderson, 1995). Cognition can be defined as the capacity to acquire, organize and use knowledge (Lidz, 1987). Basic cognitive processes that influence an infant's ability to learn and perform fine motor skills are for example attention, perception and memory (Exner & Henderson, 1995). However, to perform object related hand actions the infant uses complex cognitive functions like organizing of knowledge and as well as the use of existing knowledge to understand, plan and carry out the fine motor task (Exner & Henderson, 1995; Gallahue & Ozmun, 1995).

1.3 CEREBRAL PALSY

Cerebral palsy is caused by a damage in the developing brain, either in utero or during the first two years of life, and is a very heterogeneous diagnosis. While motor performance and posture are always affected it is also common with cognitive or perceptual impairments, epilepsy or other disabilities affecting a person's ability to perform daily activities (Rosenbaum et al., 2007). CP is classified by type based on symptoms and into subtypes based on the localization of the impairment (Surveillance of Cerebral Palsy in, 2000).

1.3.1 Prevalence and classification

CP is the most common cause of physical disability in children and the prevalence in Western countries is between 1.5 and 2.5 (Himmelmann & Uvebrant, 2018; Kragelob-Mann & Bax, 2009; Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013; Surveillance of Cerebral Palsy in, 2000). In Sweden children with CP are included in the National Cerebral Palsy surveillance program and quality register, called CPUP, which reports a prevalence of 2.6 out of 1000 in Sweden (CPUP Uppföljningsprogram för cerebrala pareser, 2018).

CP is classified into three types: spastic CP, dyskinetic CP and ataxic CP, based on the predominant neurological finding. The most common type of cerebral palsy is spastic CP which, in addition to pathologic posture and affected movement pattern, demonstrate increased tone and pathological (Surveillance of Cerebral Palsy in, 2000).

Spastic CP is further divided into sub-types based on distribution of the impairment where unilateral spastic CP implies involvement of hand, arm and leg on one side of the body

(Surveillance of Cerebral Palsy in, 2000) and constitutes the largest subgroup of CP with a prevalence of 44% in western Sweden (Himmelmann & Uvebrant, 2018).

1.3.2 Etiology

There are a large number of risk factors associated with CP, maternal risk factors are for example infections during pregnancy, epilepsy and low socioeconomic status (McIntyre et al., 2013). Risks factors that are associated with the infants are for example prematurity, low birth weight, asphyxia, neonatal seizures and infections (McIntyre et al., 2013) but the single most influential factor is birth week (Sukhov, Wu, Xing, Smith, & Gilbert, 2012). The prevalence of CP in infants born extremely preterm (<gestational week 28) is 59 out of 1000 live birth while it is 6 out of 1000 for infants born moderate to late preterm (gestational week 32-36) and only 1.2 out of 1000 in infants born full term (Himmelmann & Uvebrant, 2018).

CP is a caused by a non-progressive disturbance in the developing brain, either in utero or during the first years of childhood (Rosenbaum et al., 2007). The consequences of the lesions vary due to many factors such as timing, size and site of the lesion but also other still unknown factors (Hadders-Algra, 2014). Neuroimaging through Magnetic Resonance Imaging (MRI) is commonly used to characterize the lesion and about 86% of children with CP has an abnormal MRI. The most common origin of CP is WMI (White Matter Injury of immaturity), that is periventricular lesion in the white matter followed by cortical and deep grey matter lesions and brain maldevelopments (Himmelmann & Uvebrant, 2018; Krageloh-Mann & Horber, 2007; Towsley, Shevell, Dagenais, & Consortium, 2011).

Predominantly periventricular white matter lesions are the most common for children with unilateral CP followed by and predominantly and deep grey matter lesions while 10-15% of children with unilateral CP do not have abnormal MRI (Hadzagic-Catibusic, Avdagic, Zubcevic, & Uzicanin, 2017; Krageloh-Mann & Cans, 2009).

1.3.3 Comorbidity

Although CP implicates a movement disorder causing an activity limitation it is also usually accompanied by secondary problems such as disturbed sensation, perception, cognition, epilepsy or other (Granild-Jensen, Rackauskaite, Flachs, & Uldall, 2015; Rosenbaum et al., 2007). Many children with CP also have some degree of intellectual disability, but reported prevalence varies from 23-56% (Krageloh-Mann & Bax, 2009; Surveillance of Cerebral Palsy in, 2000). Vision impairments are present in about 50-70% of children with CP and can range from total blindness to less severe and isolated impairment like a narrowed field of view.

However visual impairments are less common and usually less severe for infants with a unilateral CP (Dufresne, Dagenais, Shevell, & Consortium, 2014; Fazzi et al., 2012; Mercuri, Guzzetta, & Cioni, 2008). Infants with CP often has an affected trunk control which also influence their ability to reach for and grasp objects and for infants with unilateral CP there is an increased involvement of the trunk when reaching for objects (Steenbergen et. al, 2000; Charles, 2008).

1.3.4 Diagnosis

CP is a clinical diagnosis that is set based on a combination of clinical signs and neurological examinations. Diagnosis is usually set between 12 and 24 months but the clearer the signs are and the earlier the child will be diagnosed (Novak et al., 2017). A diagnosis of spastic unilateral CP includes abnormal pattern of posture and movement, increased tone and pathological reflexes (Surveillance of Cerebral Palsy in, 2000).

In Sweden infants with a high risk of CP should be followed clinically by a multi-professional team with the aim to identify infants who have a suspected CP as early as possible. To provide infants and parents with the correct interventions infants should be diagnosed as early as possible but no later than at 18 months of age (Neonataleforeningen, 2015). A recent study on Danish children with CP showed that the median age for children to get the diagnosis unilateral CP is 13.5 months of age and children who were able to walk without aid were diagnosed even later (Granild-Jensen et al., 2015).

To set the clinical diagnosis cerebral palsy at an early age a combination of standardized assessment tools should be used. If there are suspicions of a diagnosis of CP but it cannot yet be confirmed, the diagnosis high risk of CP should be used and the infants should receive cerebral palsy specific interventions (Novak et al., 2017).

Parents wish to be informed about the diagnosis as soon as possible and most parents suspect that their child has some type of difficulty before the diagnosis is set (Baird, McConachie, & Scrutton, 2000). The earlier an infant is diagnosed with either CP or high risk of CP the earlier the infant, as well as the parents, can get access to the interventions and the support that is available (McIntyre, Morgan, Walker, & Novak, 2011). Early detection of infants that are at risk of CP or other neurodevelopmental disorders is of great necessity to enable intervention as early in life as possible (Cioni, Inguaggiato, & Sgandurra, 2016).

1.4 DEVELOPMENT IN HAND USE IN CHILDREN WITH UNILATERAL CP

Children with unilateral CP has one better functioning hand and one affected, or lesser functioning, hand. The manual ability of the lesser functioning hand can range from a mild impairment to an inability to use the hand, but it is more apparent in grasping than in reaching (Steenbergen et al., 2000; Arnould, Bleyenheuft, & Thonnard, 2014; Rosenbaum et al., 2007). An early sign of unilateral CP is asymmetric hand use that sometimes can be seen as early as at 3 months of age (decreased wrist movement and dissociated finger movements) (Guzzetta et al., 2010). In most cases though, the first sign is said to emerge at 4-5 months of age when infants reach for objects distinctly more often with one hand, something that is often described as early hand preference (Golomb et al., 2001; Krageloh-Mann & Bax, 2009). For infants the lesser functioning hand is more often fisted (Krageloh-Mann & Bax, 2009) and when they do reach for objects, with the affected hand, the reach is slower and less precise than that of a typically developing infant (Charles, 2008).

Infants with clinical signs of unilateral CP improves their use of the affected hand during the first year of life and the development follows three different trajectories. All three groups improved their use of the hands but for the low functioning group, that did not use active grasp, development was slow and seemed to plateau at nine months of age. The moderate functioning group and high functioning group both improved their hand use continuously during the first year of life but at a faster pace for infants in the high functioning group (Sakzewski, Sicola, Verhage, Sgandurra, & Eliasson, 2018).

1.4.1 Fine motor interventions for children with CP

For older children with CP interventions to improve fine motor skills are common in every day clinical practice, even though there is a gap between what is done in clinical practice and what has been shown improve fine motor function in research (Novak et al., 2013). Two of the more common interventions aimed at improving fine motor skills for children with unilateral CP are Constraint Induced Movement (CIMT) and bimanual training and there is solid evidence for the both of them (Novak et al., 2017) but no clear consensus as to which intervention is more effective (Klepper, Clayton Krasinski, Gilb, & Khalil, 2017; Tervahauta, Girolami, & Oberg, 2017).

1.4.2 Fine motor interventions for infants with CP

The rapid neurodevelopment during the first year of life provides a unique window of opportunity for a successful outcome of intervention aimed at improving motor function (Basu

et al., 2014; Hadders-Algra, 2014). Motor interventions for infants at risk of developing CP should preferably be intense and activity-based (Arnould et al., 2014; Sakzewski, Ziviani, & Boyd, 2014) and a model where the parents are coached to perform the training in the home seems to be a good model (Hadders-Algra, 2011). Studies of early interventions aimed at improving arm and hand function in infants with brain lesions have shown promising results (Cioni et al., 2016; Sakzewski et al., 2014) and a number of ongoing studies are discussed below.

CIMT is an upper extremity training where the better functioning or unaffected hand is constrained to encourage the use of the affected or poorer functioning hand during intensive training (Gordon, 2011). CIMT has recently been modified to be feasible for children under the age of 12 months. The new adjusted version is called Baby-CIMT, where both the dosage, the restraint and the task has been modified to suit infants (Eliasson, Sjostrand, Ek, Krumlinde-Sundholm, & Tedroff, 2014). Baby-CIMT has shown positive results on bimanual hand function for infants with an asymmetric hand use (Eliasson et al., 2018). A retrospective study shows that children with unilateral CP that have received Baby-CIMT before the age of 12 months are six times more likely to have a high functional level regarding hand function at two year of age than children that has not received the treatment (Nordstrand, Holmefur, Kits, & Eliasson, 2015). There is also an ongoing randomized comparison trial comparing baby-CIMT to baby BIM, that is infant-friendly bimanual therapy, where the only thing that differs between the groups is the type of training they receive while other factors such as dosage, setting and support from therapists are the same (Boyd et al., 2017).

Early Therapy in Perinatal Stroke (eTIPS) is a parent-delivered intervention during the first six months for infants that has had a perinatal stroke. The goal is to achieve improved motor function in the affected side of the body by enforcing motor activity and visual and sensory stimulation as a part of daily life (Basu, Pearce, Baggaley, Watson, & Rapley, 2017). The eTIPS is feasible both for parents and therapists and the effect of eTIPS for infants with perinatal stroke will be evaluated in a Randomized Controlled Trial (RCT) (Basu et al., 2018).

Action observation therapy is based on the concept of mirror neurons and for this intervention active training of hand and arm movements is combined with observation of hand actions. Studies have shown promising results for adults with unilateral stroke as well as for children with unilateral cerebral palsy (Burzi, Tealdi, Boyd, & Guzzetta, 2016). An RCT is now ongoing to evaluate the effect of action observation therapy on hand and arm movements for infants with unilateral brain lesions (Guzzetta et al., 2013).

Studies have shown that the bigger brain lesion an infant has, the lesser the chance is to benefit from interventions (Hadders-Algra, 2014). This presents a problem, where the infants that least will benefit from training receive the diagnosis early while those infants that would benefit the most from intervention run a risk of receiving their diagnosis too late, when the crucial window of opportunity perhaps already has closed. This shows a need for sensitive tests that can distinguish infants that run a risk of developing CP at an earlier age.

1.5 ASSESSMENTS

When using an assessment, the purpose of the assessment needs to guide the choice of instrument (Krumlinde-Sundholm, 2008). So if we intend to evaluate object related use of hands there is a need for an assessment that has a separate sub scale for fine motor performance.

1.5.1 Motor assessments for infants

However, to identify infants with clinical signs of unilateral CP the assessment also needs to provide a measure for each hand separately in order to have an outcome demonstrating a possible side difference between the hands (Krumlinde-Sundholm et al., 2015).

There are a number of available assessment tools for infants that include aspects of upper limb motor proficiency in infants but that lacks a separate sub scale for fine motor skills (Krumlinde-Sundholm et al., 2015). One example of that is the Infant Motor Profile (IMP) which is an observation based assessment tool for infants between 3 and 18 months. The assessment was constructed to identify infants with developmental motor disorders and contains five sub scales but does not have a separate sub scale for fine motor skills (Heineman, Bos, & Hadders-Algra, 2008). Other assessment tools that include fine motor items but that does not have a separate fine motor sub scale are the Movement Assessment for Infants (MAI) (Chandler & Andrews, 1980) and the Toddler and Infant Motor Evaluation (TIME) (Tieman, Palisano, & Sutlive, 2005).

Some assessment tools that are used for infants do have a separate sub scale for fine motor performance. One example of this is the Peabody Developmental Motor Scale (PDMS) that is a widely used observation based assessment tool. PDMS contains two different fine motor domains, Grasping and Visual-motor integration. PDMS can be used from birth to five years of age and for infants 12 months old and younger there are 19 items in the domain for Grasping and 31 items to measure Visual-motor integration. Five of the items are bimanual while the rest of the items are unimanual and can be performed using the preferred hand (Folio & Fewell, 2000). Another assessment tool with a separate sub scale is the Bayley Scale of Infant and

Toddler Development (BSITD). The BSITD is constructed to detect infants with a delayed motor development and includes both bimanual and unimanual items. However as for the PDMS the BSITD measures the preferred hand only and will not be able to detect a possible side difference between the hands (Bayley, 2006). In using assessment tools, like the ones mentioned above, with a majority of items measuring preferred hand infant with a unilateral CP may performed within the norms since they can use their unaffected hand (Novak et al., 2017).

The Structured Observation of Motor Performance (SOMP-I) is an observation based motor assessment to be used at set ages during the first year of life. The SOMP-I evaluates different aspects of motor development as well as quality of movements. The main focus of SOMP-I is gross motor development but one scale includes aspects of fine motor function which are measured in the supine position (Persson & Stromberg, 1995). SOMP-I might be used to detect infants with CP even though the low specificity, at highest 72% at 6 months of age, would lead to infants receiving a diagnosis of CP that is later removed (C. Montgomery, Johansen, Lucas, Stromberg, & Persson, 2017).

The Developmental Assessment of Young Children second edition (DAYC-2) is a criterion and norm referenced assessment meant to detect children with developmental delay. The DAYC-2 has five domains whereof the “Physical development-domain” contains two sub domains, one for gross motor and one for fine motor. The items are scored 1 (passed) or 0 (not passed) and the fine motor sub domain contains 33 items with a floor effect at 9 months (Judith & Maddox, 2014).

1.5.2 Assessments of hand use for children with unilateral CP

For older children (aged 18 months-18 years) with unilateral CP the Assisting Hand assessment (AHA) is a widely used assessment that measures how effectively the affected hand is used in bimanual activities (Holmefur & Krumlinde-Sundholm, 2016; Wallen, 2016). The Mini Assisting Hand Assessment (Mini AHA) is development of the AHA and is valid to measure use of the affected hand in for children 8-18 months with unilateral CP (Greaves, Imms, Dodd, & Krumlinde-Sundholm, 2013).

However, for infants that have not yet been diagnosed with unilateral CP but demonstrate asymmetric hand use there was until recently no suitable assessment. For these infants with clinical signs of unilateral CP it is important to be able to measure each hand separately in order

to have an outcome demonstrating a possible side difference between the hands (Krumlinde-Sundholm et al., 2015).

Since the start of this work to develop a new assessment tool the Grasp and Reach Assessment of Brisbane (GRAB) has been developed. The GRAB is a criterion referenced assessment tool that is intended to be used in research. The GRAB was developed to detect an asymmetry in upper limb movements in the reaching and grasping of infants that are at risk of developing unilateral CP. The GRAB can be used at set ages (14, 16 and 18 weeks' gestational age) and consists of a structured play session where three specific toys are presented to the infants. The toys are presented to the infant at shoulder height and in mid line from behind a screen. The GRAB consists of seven unimanual and two bimanual behaviors that are scored. The assessment consists of six toy presentations and each toy is presented for 30 seconds followed by a 30 seconds brake. The assessment is filmed and edited, so that each toy presentation is a separate video-clip, before scoring (Perez, 2016; Perez et al., 2016).

1.5.3 Criterion referenced and norm referenced tests

Tests can be either criterion-referenced, norm-referenced or both. In a criterion-referenced test you investigate whether the infant meet pre-determined criteria within one or more well-defined domain(s). Criterion referenced tests are often used to evaluate change since the result is not set in relation to the age of the infant but rather to how large part of the tested domain the infant has mastered (Krumlinde-Sundholm, 2008).

Norm referenced tests provide age norms that represent typical behavior of the measured skills and the infants' performance can be compared to the average performance of a sample of typically developed age-matched peers (Cohen, Swerdlik, & Sturman, 2013; Mitrushina & ebrary Inc., 2005; P. C. Montgomery & Connolly, 1987). The results are commonly reported as standard deviation-based scores to indicate how far from the mean score of the normative sample the infant's performance is (Weiss, 2016). Standardized tests used in pediatric clinical settings are most often norm referenced (Hanna et al., 2007; P. C. Montgomery & Connolly, 1987). For motor assessments for infants, age norms are considered especially important due to the rapid pace of development during the first year of life (Hadders-Algra, 2014; Mitrushina & ebrary Inc., 2005).

1.6 PSYCHOMETRIC PROPERTIES

The psychometric properties, or measurement properties, of a test is the evidence of the statistical strength or weakness of the test. Psychometric properties relate to the quality of a

test and validity and reliability are the most common measurement properties (Souza, Alexandre, & Guirardello, 2017).

1.6.1 Test theory (modern and classical)

Classical Test Theory (CTT), also called True Score Theory, is used for methods that are in fact traditional rather than classical, and includes our traditional ways of evaluating psychometric properties of an assessment. It is based on the idea that each test has a true score and the score you actually get as a result of an assessment, the observed score (X), consists of the true score (T) and the error (E) that is $X=T+E$. (DeVellis, 2006; Streiner, Norman, & Cairney, 2015).

If all error could be deducted only the true score would remain, unfortunately both the true score and the error are unobservable but the goal of classical test theory is to improve the precision of a test by minimizing the error (Kimberlin & Winterstein, 2008). Classical test theory acknowledges the fact that each item has individual random error but the main emphasis is on the whole scale. There are different ways to attempt to estimate the random error of the scale, for example test-retest and interrater reliability (DeVellis, 2006).

Item response theory (IRT), also called modern test theory, is a theory about how to evaluate the properties of the results of an assessment. The theory behind IRT is that the test subject has a latent ability and the goal is to assess that underlying trait. In IRT the focus is on the item level (Streiner et al., 2015). In IRT the items are arranged in order of difficulty and the persons are arranged in order of competence in the same scale (Downing, 2003). The most widely used model within IRT, is the Rasch measurement model analysis, or one parameter item response model, which has been widely used and become a preferred method for creating new tests and for evaluation existing outcome scales (Tennant & Conaghan, 2007).

1.6.2 Validity

Validity has been defined in “Standards for educational and psychological testing” (Standards), page 9, as “the degree to which evidence and theory support the interpretation of test scores entailed by proposed uses of tests” (American Educational Research Association., American Psychological Association., National Council on Measurement in Education., & Joint Committee on Standards for Educational and Psychological Testing (U.S.), 1999).

As has been emphasized it is not the test itself that is valid but rather the result of the test when performed as described and for a specific target group (American Educational Research

Association. et al., 1999) To use the test for a new population validity has to be evaluated for the new target group (Streiner et al., 2015).

Traditionally there have been three main types of validity, the three C's, Content validity, Construct validity and Criterion validity (Streiner et al., 2015).

Content validity refers to whether the test collects all relevant information necessary to measure the underlying trait we intend to measure. This can best be examined by experts in the field (Salkind, 2014; Streiner et al., 2015). Criterion validity relates to how well the scores of the test agree with the performance for what we want to measure. This can be examined by calculating the correlation of the results of a new assessment with the result of an existing assessment, measuring the same trait we intend to measure. This is used when there is an existing assessment for what we want to measure, a 'golden standard' that we for some reason, for example economic, want to replace (Streiner et al., 2015; Vet, 2011). Construct validity relates to whether the two tests measure the same underlying construct or trait (Streiner et al., 2015).

More recently there has been a shift in the concept of validity and there is no longer considered to be different separate types of validity but validity is considered to be a unitary concept (American Educational Research Association. et al., 1999; Goodwin, 2002). There is now according to Standards five different sources of evidence of validity (see table 1)

Table 1. Different sources of evidence of validity and how they relate to the earlier concepts of validity (Goodwin, 2002).

1974	1999
Content validity	Evidence based on test content
Criterion validity	Evidence based on relation to other variables
Construct validity	Evidence based on internal structure
	Evidence based on response processes
	Evidence based on the consequences of testing

Evidence based on test content was earlier called content validity and relates to how well the items of the test represents the trait it is intended to measure. This can be evaluated by a group of experts within the field. We want to know whether all aspects of the trait are measured or if there should be more items. Or maybe if we have items that are irrelevant to the trait. (American Educational Research Association. et al., 1999).

Evidence based on relation to other variables. This is a way to investigate the relation between the scores of an assessment and external variables. This can be examined by investigating the correlation of the results of two assessments that claim to measure the same trait, for example if there are two different methods of measuring the same construct (Goodwin, 2002). This can be used when there is an existing assessment for what we want to measure, a 'golden standard' that we for some reason, for example economic, want to replace (Streiner et al., 2015).

Evidence based on internal structure can be evaluated by analyzing whether the items in a test forms one unidimensional construct. This can be investigated through factor analysis, principal component analysis or item response theory, for example Rasch measurement model analysis (Streiner et al., 2015). Another way to investigate this is to investigate Differential Item Functioning (DIF) (Goodwin, 2002) DIF is present if infants that have the same ability but belongs to different sub groups, based on for example gender or nationality, responds significantly different on individual items (Bond & Fox, 2015; Goodwin, 2002).

Evidence based on response processes is evaluated by analyzing the response pattern or individual responses of the test takers by, for example interviews. However, there is also a need to investigate the response process of the person scoring the assessment. Whether the scorers are applying the items as intended or if the scoring process is influenced by irrelevant factors (American Educational Research Association. et al., 1999)

Evidence based on the consequences of testing is evaluated by investigating whether there are expected or unexpected benefits or disadvantages to using an assessment (American Educational Research Association. et al., 1999).

1.6.3 Validity evaluated by Rasch measurement model analysis

The Rasch measurement model, or the one parameter item response model, is widely used to evaluate the psychometric properties of an assessment tool (A. G. Fisher, 1993). It can also be used when a new test is constructed to investigate unidimensionality of the scale and fit of the items (Tennant & Conaghan, 2007).

The fundamental thought in the Rasch measurement model is that if the items measure only one construct or trait, all items can be placed in order from the easiest to the hardest while all persons can be placed in order from least competent (within the measured trait) to the most competent (within the measured trait) on the same line (Bond & Fox, 2015).

There are two assertions underlying the Rasch model: All persons are more likely to pass an easy item than a difficult one. All items are more likely to be passed by persons of high ability than by persons of low ability (Bond & Fox, 2015; Wright & Stone, 1979).

By use of the Rasch model measurement analysis data is analyzed to investigate how well it fits the two assertions. If the data fit the model assertions the assessment tool measures one and the same underlying trait (Bond & Fox, 2015). By use of the Rasch model measurement analysis data is analyzed to investigate how well it fits the two assertions. If the data fit the model assertions the assessment tool measures one and the same underlying trait (Bond & Fox, 2015).

1.6.4 Reliability

Reliability refers to how exact the result of a test is, that is how much random error is in the result and how close the score of the assessment is to the true score (Mokkink et al., 2010). It is the consistency of a score across time or between raters that is investigated through investigating the level of agreement between two assessments or scorings (Vet, 2011).

Different types of reliability are for example intra rater reliability, inter rater reliability, test-retest reliability and internal consistency. Intra rater reliability relates to the rater's self-consistency in scoring, that is if the rater scores the same infant twice, from the same test situation (for example a video recorded session), will the scores be consistent and show a small variation in its scores (Balakrishnan, 2014). The inter rater reliability refers to whether two raters score the same assessment with high agreement while the test-retest reliability, is investigating whether the infant demonstrates the same behavior if the test is repeated at a different time point when no change is expected (Streiner et al., 2015).

Another aspect of reliability is internal consistency which refers to to what degree each item of a scale measures the same underlying trait, do the items all correlate to the result of a test (Pallant, 2010).

Like with validity there really are no reliable tests but rather reliable scores or the reliability of a test as applied to a specific test situation (Streiner et al., 2015).

2 RATIONALE FOR THIS THESIS

Early intervention for infants with signs of unilateral CP is recommended in the literature but the lack of appropriate assessment tools has limited the possibility to identify eligible infants and to evaluate the outcome of the interventions. An assessment tool that could quantify the ability level of each hand separately, provide a measure of asymmetry in hand use and provide a measure for the overall bilateral hand use for infants would make that possible. An assessment with those properties would make it possible to: (i) properly describe hand use in infants (ii) distinguish infants demonstrating signs of unilateral CP that would benefit from early intervention and (iii) evaluate a change in hand use and thereby properly evaluate early intervention.

The need for this thesis is there for threefold (i) clinicians need to be able to identify infants eligible for early interventions and correctly report the outcomes of the interventions (ii) researchers need to be able to properly evaluate the effects of early intervention (iii) parents request (Baird et al., 2000) and deserve information at an earlier stage about the abilities of their infants as well as the difficulties that may lay ahead of them.

3 AIMS OF THE THESIS

The over-all aim of this thesis was to develop an assessment tool measuring bilateral goal-oriented hand use in infants 3-12 months of age, to investigate its psychometric properties for infants that shows signs of unilateral CP as well as to create normative reference values.

The thesis contains three Studies with the following specific aims:

Study I:

To describe the development of the Hand Assessment for Infants (HAI) and to investigate aspects of internal scale validity and reliability of the HAI for infants with signs of unilateral CP, using Rasch measurement model analysis.

Study II:

To create normative reference values for the HAI by assessing typically developing infants between 3 and 10 months of age.

Study III:

To evaluate the test-retest and interrater reliability of the sum score and the measure of the HAI for infants with signs of unilateral CP.

4 METHODS

4.1 PARTICIPANTS

Infants with asymmetric hand use and infants with a typical development were recruited in Sweden, Italy and the Netherlands for the Studies (table 1). All participating infants were between 3 and 12 months old.

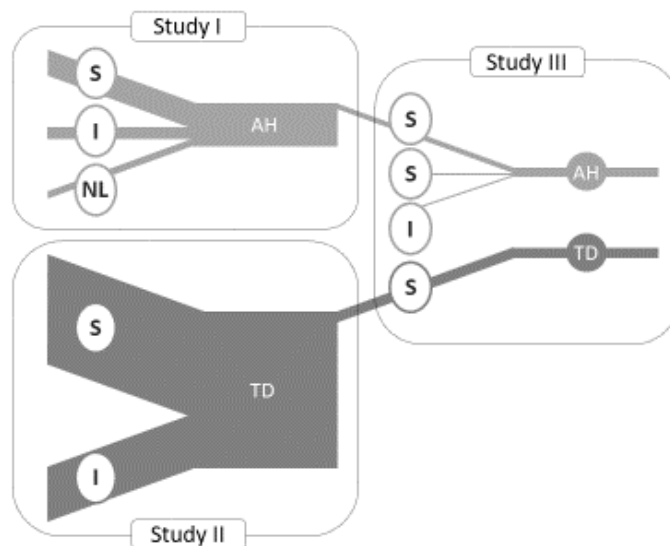
Table 2. Participants included in Studies I, II and III.

	Study I				Study II			Study III		
	Total	Swe	Ita	NL	Total	Swe	Ita	Total	Swe	Ita
Total	124	72	32	20	451	297	148	55	50	5
AH	124	72	32	20				25	20	5
TD					451	297	148	30	30	

Swe Swedish, *Ita* Italian, *NL* Dutch, *AH* Asymmetric hand use, *TD* Typically developing

Fifteen of the Swedish infants with an asymmetric hand use in Study I also participated in Study III and 30 of the typically developing infants from Sweden in Study II also participated in Study III (figure 1).

Figure 1. Flow chart of infants participating in more than one study.



S Swedish, *I* Italian, *NL* Dutch, *AH* Asymmetric hand use, *TD* Typically developing

4.1.1 Assessments

For the first two Studies in this thesis some infants were assessed using the HAI more than once and the assessments were considered as individual data points. For Study I 32 of the infants were assessed twice and for Study II 16 of the infants were assessed more than once. In Study III all infants were assessed twice within seven days as a part of the test-retest study design. The number of assessments in each study is shown in table 2.

Table 3. Number of assessments in Studies I, II and III.

	Study I				Study II			Study III		
	Total	Swe	Ita	NL	Total	Swe	Ita	Total	Swe	Ita
Total	156	94	38	24	489	331	158	110	100	10
AH	156	94	38	24				50	20	10
TD					489	331	158	60	60	

Swe Swedish, *Ita* Italian, *NL* Dutch, *AH* Asymmetric hand use, *TD* Typically developing

4.1.2 Study I

In Study I 156 assessments of 124 infants with asymmetric hand use were included. The sample included 20 Dutch infants, 32 Italian infants and 72 Swedish infants. Some children were assessed twice and in total 156 assessments were included, 94 Swedish, 38 Italian and 24 Dutch.

4.1.3 Study II

In Study II 451 typically developing infants between 3 and 10 months of age participated. Sixteen of the infants were assessed more than once ((n children: n assessments), 11:2, 1:4, 1:6, 2:7, 1:8) which resulted in a total of 489 assessments. Most of the assessments were collected in Sweden (331 assessments of 297 infants) while a smaller part was collected in Italy (158 assessments of 148 infants).

4.1.4 Study III

All of the 30 typically developing infants included in Study III were recruited in Sweden and were also a part of the sample in Study III. Twenty of the infants with an asymmetric hand use were recruited in Sweden and all but five were also a part of Study I. The five infants with asymmetric hand use that were recruited in Italy was not a part of the sample in Study I. All infants were assessed with the HAI twice during one week. The first assessment was scored by

two scorers to investigate interrater reliability and both assessments of an infant was scored by the same scorer to investigate test-retest reliability.

4.1.5 Recruitment process

Infants with an asymmetric hand use were recruited in three countries, Sweden, Italy and the Netherlands. They were all identified by clinically experienced occupational therapists, physical therapists or medical doctors as being at risk of unilateral CP and the asymmetric hand use was confirmed through assessment with the HAI.

Infants with a typical development were recruited in Sweden and Italy. The sample is partly a convenience sample where colleagues, friends and acquaintances were asked if their children would participate. The majority of the Italian sample of typically developing infants were recruited through leaflets at a pediatrician's office in Livorno where parents were asked to participate. The majority of the Swedish sample was recruited at open play schools and child health care centers in greater Stockholm where all parents present were asked by a research assistant if their infant could participate. All of the typically developing infants were reported by the parents as having no known neurological events up to the date of the assessment as well as no known deviation to typical development.

4.1.6 Data collection and assessments

All data for this thesis, except the descriptive data of the infants and parents, were collected through video recordings of the semi-structured play session of HAI. The video recordings were performed at different places: in the homes of the infants, in research facilities at Karolinska Institutet in Stockholm, Stella Maris in Pisa or Utrecht, or on location at the open play schools or child health care centers. The films were then scored using the research version of HAI and re-scored when changes were made to the scoring criteria. The video recordings have made it possible to re-score the same assessments a large number of times.

4.2 STATISTICAL ANALYSES

The statistical analyses were performed using IBM SPSS software (IBM Corporation, Armonk, NY) except the Rasch model measurement analysis in Study I which was performed using WINSTEPS software version 3.92.1 (John M Linacre, 2015). The statistical significance was set to $\alpha=0.05$. An overview of the methods of analyses can be seen in table 3.

Table 4. Overview of statistical methods used in Studies I, II and III.

Statistical methods	Study I	Study II	Study III
Bland-Altman plot			x
Descriptive statistics	x	x	x
Independent samples t-test		x	x
Intraclass Correlation Coefficient			x
Kruskal Wallis	x		
Mann- Whitney U	x		
Paired samples t-test		x	x
Pearson correlation coefficient		x	
Rasch measurement model analysis	x		
Smallest Detectable Difference			x
Spearman's rank correlation coefficient	x		
Standard Error of Measurement			x

4.2.1 Descriptive statistics

Descriptive statistics are used to describe the different characteristics of a sample, like for example age and sex. Examples of descriptive statistics are frequency, minimum value, maximum value, mean and Standard deviation (SD) (Pallant, 2010; Salkind, 2014).

4.2.2 Measures of validity

4.2.2.1 Rasch measurement model analysis

The Rasch measurement model analysis was used to investigate aspects of internal scale validity and reliability of the HAI.

The polytomous model was chosen since there are three scale-steps for each item and since the thresholds between scale steps was expected to differ the Masters partial credit model was chosen (Masters, 1982; Tennant & Conaghan, 2007).

Person and item fit was evaluated by inspecting infit MnSq and z-values. An infit MnSq of ≤ 1.4 in combination with a z-value of < 2 was considered acceptable goodness of fit for both items and persons. A total number of 95% person and item fit is considered acceptable (Chien & Bond, 2009).

Unidimensionality is investigated through Principal Component Analysis (PCA) of the standardized residuals. The principal component, or first dimension, should explain $> 60\%$ of the result and the second dimension should explain $\leq 5\%$ of the result. Otherwise the assessment

could have more than one dimension, that is that it could measure more than one trait, or skill (W. Fisher, 2007).

Separation was calculated to investigate how many statistically distinguishable ability levels, or strata, that existed in the sample. An assessment tool needs to have a strata greater than 2 to separate people into at least two levels of ability (W. Fisher, 2007).

The Rasch measure model analysis should also be used when the result on several items are added to a total score (Tennant & Conaghan, 2007).

The Rasch measure model analysis transforms raw scores from ordinal scales to interval data, called logits, through a logarithmic transformation of the probability of success. The use of logits offers the possibility to compare changes equally over the full range of the scale. In interval data, all scale steps are equal in size and results can therefore be compared across levels (Bond & Fox, 2015).

Rasch measurement model analysis sorts the persons in order of capability or competence, ordering them from less competent to more competent. The items are ordered from easiest to most difficult along a measurement line, thus creating a hierarchy order of the items. This hierarchy can be considered to describe the order in which the abilities develop (Bond & Fox, 2015).

4.2.3 Measures of reliability

4.2.3.1 Intraclass Correlation Coefficient

The Intraclass Correlation Coefficient (ICC) is a measure of the strength of the agreement between two results. The ICC has a value between 1 and 0 where an ICC close to 1 is considered large and there is high reliability. The higher the ICC the lower the error (Streiner et al., 2015).

Two-way random effects model single rater/measurement of absolute agreement (or ICC 2.1) was used to investigate the reliability of the HAI in Study III. Two-way random effect model is most often used to investigate the reliability of clinical assessments. This model is used if the raters are chosen from a population of raters who have an extensive knowledge of the assessment. The single measurement or single rater type is used to compare the results of two single measures rather than the mean of several assessments (Koo & Li, 2016). Absolute agreement is used if you

There is no real consensus of how ICC should be interpreted (Streiner et al., 2015). Some claim that an ICC value between 0.4 and 0.75 could be considered as fair to good reliability (Lexell & Downham, 2005) while others claim that an ICC value of 0.75 should be considered the minimal for an assessment to be considered useful at all (Streiner et al., 2015). However most agree that an ICC above 0.9 could be considered excellent (Koo & Li, 2016; Lexell & Downham, 2005) (Lexell & Downham, 2005) and indicates that the scale can be used both in research and in clinical work (Streiner et al., 2015). For this work the information from Koo et al was used and ICC was interpreted as follows: poor (<0.5), moderate (0.5-0.75), good (0.75-0.9) and excellent (>0.9) (Koo & Li, 2016).

4.2.3.2 *Standard error of Measurement*

Standard error of measurement (SEM) is an absolute index of reliability. SEM is an index that can be used to define the difference in score that is needed for the change to be considered real (Weir, 2005).

The SEM allows us to quantify to which extent a test provides accurate scores. Low SEM indicates high levels of score-accuracy. The SEM is calculated $SEM = SD \sqrt{1-r}$ where the SD is calculated based on the first of the two assessments of test-retest and the reliability measure (r) is the ICC of the score that is investigated. The SEM is expressed in the same unit as the measurement (Streiner et al., 2015).

4.2.3.3 *Smallest Detectable Differences*

The Smallest Detectable Difference (SDD) is a change that is bigger than the measurement error. It is the smallest difference in points a person would have to improve for the assessor to know that there is a true change of the underlying trait that is measured. SDD is calculated $SDD = SEM \times 1.96 \times \sqrt{2}$ (Lexell & Downham, 2005).

4.2.3.4 *Bland Altman plot*

A Bland-Altman plot is a method that visualizes the agreement between assessors or assessments. The Bland-Altman plot is a scatterplot where the mean of two assessments is plotted on the Y-axis while the difference between the two assessments is plotted on the X-axis. In the plot the mean of all assessments as well as the upper and lower limit is marked with lines (Bland & Altman, 1986). The plot is visually inspected to examine whether there is a systematic difference between two assessments (Streiner et al., 2015). Bland Altman plots are primarily used to compare two different methods of measurement but can be used to analyze

the repeatability of a measure as well as the possible existence of a difference between two raters (Bland & Altman, 1986).

4.2.3.5 *Cronbach's alpha*

Cronbach's alpha can vary from 0 to 1 and is a measure of internal consistency reliability, that is it is a measure of to what the degree the items of an assessment measure the same underlying attribute (Campbell, Walters, & Machin, 2007). However, Cronbach's alpha is also influenced by the amount of items in the assessment and a low Cronbach's alpha could indicate that the assessment measures more than one trait or that there are too few items while a Cronbach's alpha close to 1 could mean that the assessment includes redundant items (Tavakol & Dennick, 2011).

4.2.4 **Comparing groups**

4.2.4.1 *T-tests*

The t-test is used to determine whether there is a statistically significant difference between two groups (Pallant, 2010). There are two types of T-tests that both compare the mean score of two groups, the independent t-test and the paired t-test. The independent t-test is used when you want to compare means of the result of two different groups to investigate for example whether there is a difference between men and women (Skaik, 2015). The paired t-test should be used to compare two means of groups that are in some way related, for example to compare the results at base line and after an intervention (Pallant, 2010), but can also be used to compare two results of the same subject for example of compare the right and left hand (Skaik, 2015).

4.2.4.2 *Mann-Whitney U*

The Mann-Whitney U is a non-parametric method that is used to compare the results of two independent groups on a continuous measure (Campbell et al., 2007; Pallant, 2010). The Mann-Whitney U is chosen over the independent samples t-test if data is not normally distributed (Pallant, 2010).

4.2.4.3 *Kruskal Wallis*

The Kruskal Wallis is used to compare the result of three or more independent groups (Campbell et al., 2007; Pallant, 2010).

4.2.5 Exploring relationships

Investigation of correlation is performed to describe the linear relationship between two variables (Pallant, 2010).

4.2.5.1 Pearson correlation coefficient

The Pearson correlation coefficient is used to investigate if there is a correlation between two continuous variables. The correlation coefficient can range from -1 to +1 (Campbell et al., 2007). A correlation of 0 indicates no relationship between variables while a correlation of 1 indicates a perfect correlation (Pallant, 2010). A correlation between 0.5 and 1.0 can be considered large (J. Cohen, 1988).

4.2.5.2 Spearman's rank correlation coefficient

The Spearman's rank correlation coefficient is used to investigate the relationship between variables if data is not normally distributed (Campbell et al., 2007).

5 ETHICAL CONSIDDERATIONS

Ethical approval for the Swedish part of the included studies was obtained from the Regional Ethical Review board (2007/1470-31, 2008/1:2, 2009/1100-32, 2016/1635-32, 2018/1329-32). The Italian part of these studies have the ethical approval of Comitato Etico (Prot no3/2011, Comitato Etico) while the Medisch Ethische Toetsingscommissie, found that no ethical approval was needed for the Dutch part of these Studies (WAG/th/14/038370). The Studies were all conducted in accordance with the Declaration of Helsinki.

All parents have signed a consent form for their infants to participate in the studies. Even so, it is important to make your own ethical considerations and since the children in our studies are so young we needed to consider both the infant and the parent point of view.

The infants are all subjected to assessments using HAI, that is a filmed semi-structured play session. One of the key elements when using HAI is that the infant is active and curious of exploring the toys. Therefore, it is essential that the infant is in a good mood and comfortable with the situation to be able to participate in the assessment. All assessors have been instructed to take a break if the infant gets sad or seems to not enjoy it. This has rarely been a problem since infants generally seem to enjoy the play session, investigating a new set of toys.

For these studies we have had an ongoing discussion of how the parents could experience the assessment situation. For the parents of the typically developed infants it was not expected to be a problem since it was primarily a chance for them to see what their infant really was capable of doing, an assumption that was confirmed throughout our work. For the parents of the infants with an asymmetric hand use, on the other hand, it was an opportunity for them to see and sometimes experience for the first time the difficulties their infant might experience. However, all parents had already seen some medical professional on the basis of the asymmetric hand use of the infant. Most often the results of the HAI only confirmed the parents' suspicions, that there was something "wrong" none the less it was considered extra important to be aware of how to present the results to the parents.

6 RESULTS

6.1 THE FINAL VERSION OF THE HAI (STUDY I)

The work behind the HAI is extensive and stretches for over a decade, includes an international collaboration between Stella Maris in Pisa and Karolinska Institutet and involves infants from Sweden, Italy and the Netherlands. The result of this is the assessment tool the Hand Assessment for Infants that measures object related hand actions in infants between 3 and 12 months of age.

6.1.1 Items and outcome

The final version of the HAI (figure 2) consists of 17 items, twelve unimanual items and five bimanual items. For the unimanual items the left and right hand are scored separately. That means they measure actions that the infant does using one hand for example initiation of hand use towards objects in midline or pre-positioning of the arm and hand to grasp the object with a well-adjusted grasp. The result of each hand is summed to the Each Hand Sum score (EaHS) which is an ordinal scale ranging from 0-24 points raw score. This means that a possible asymmetry is demonstrated in a difference in raw scores between the EaHS of the hands but it is also presented as an asymmetry index (calculated $1 - \frac{\text{EaHS of the lesser functioning hand}}{\text{EaHS of the better functioning hand}}$) where a greater percentage indicates a greater asymmetry between the hands.

Five of the items are bimanual describing actions where the infant does use both hands together for example bimanually holding one object or transferring an object forth and back between the hands. The sum of all the items is called the Both Hands sum score (BoHS) and ranges between 0 and 58 raw scores, this sum gives an over-all score of how well the infant performs in bimanual object-related hand actions. In the Rasch measure model analysis in Study I the BoHS was transformed through a logarithmic transformation of the probability of success to a logit based interval scale called Both Hands Measure (BoHM). The BoHM ranges from 0-100 HAI-units and is the main outcome of the HAI.

One item is placed outside the scale and is not included in the sum score. This item is “Grasps when restrained” where you assess how well the infant can use the lesser functioning hand when the better functioning hand is restrained by for example the assessors hand. This item is not included in the sum score since it does not measure the underlying idea/construct of the HAI, to measure typical behavior, but kept on the score form since it provides information that is important for clinical work.


		Hand Assessment for Infants (HAI) β-version 1.1			
	Name		Diagnose		
	Date of exam.		Conductor		
	Date of birth		Assessor and date		Linda Ek
	Gest week		Pred. Birth		1900-01-00
	Age	0 years	Corr Mo:		0
	Corr. Age	0 years	Corr We:		0
	Item	Comments right hand	R	L	Comments left hand
Initiation	1. Initiates use for objects on the side				
	2. Initiates use for objects in midline				
Holding	3. Holds for a while				
	4. Quality of holding				
Grasping	5. Grasps from an easy position				
	6. Object location when grasping				
	7. Adjusts arm/hand orientation				
Movements	8. Moves fingers				
	9. Moves forearm				
	10. Moves upper arm				
	11. Quality of movements				
Use	12. Amount of hand use				
		Each hand separately sum score (0-24)	0	0	Percentage difference between hands (best-worse)/best: #####
Bimanual use	13. Bimanual object contact				
	14. Bimanual holding of object				
	15. Transfers between hands				
	16. Transfers in a sequence				
	17. Bimanual manipulation				
0		Both hands Sum score (range 0-58)	0		
	HAI	Both Hands Measure, HAI-units (0-100)	#SAKNAS!		
	Grasps when restrained*				
* rated beyond the scale, only if the infant does not even grasp from an easy position					

Figure 2. Score sheet of the HAI

6.1.2 Scoring

The play session is video recorded to enable for the assessor to fully concentrate on the play session. The video recording also provides the scorer the opportunity to focus on the scoring.

The scoring takes 30 to 45 minutes and it is important to carefully read the manual while you are scoring.

6.1.3 Certification procedure

To become a certified scorer, the therapist must take a two-day course to learn the assessment- and scoring procedure of the HAI. Then the therapist has to score two provided play sessions, collect their own kit of toys, make two assessments of their own and score them. All these steps must be approved before the therapist is certified and can use the HAI in their research or clinical work. To this date (2019-05-01) 20 HAI-courses has been held, in Sweden, Italy, England, Australia, Mexico and in the US and there are now almost 200 certified HAI scorers worldwide.

The certification process is similar to the one used for many years with the AHA and the Mini-AHA.

6.2 VALIDITY OF THE HAI (STUDY I)

For the Rasch measure model analysis for Study I 156 HAI-assessments of 124 infants, aged 3-12 months with asymmetric hand use were compiled. Data was collected in Sweden (72 assessments), Italy (32 assessments) and the Netherlands (20 assessments).

Rasch measurement model analysis was used in Study I to investigate aspects of internal scale validity and reliability. Spearmans Rho was used to investigate the relationship between age in months and the EaHS of the better functioning hand, the EaHS of the lesser functioning hand as well as the BoHM. Mann–Whitney U test and Kruskal–Wallis tests were used to investigate whether there were any significant differences in results between groups based on sex, affected side, country of scoring, country of origin, and between lesser- and better functioning hands.

There were no disordered thresholds and all of the persons and items in the analysis of the final version of the HAI showed an acceptable goodness of fit. The analysis showed that the HAI measures one construct as demonstrated by the principal component analysis showing that the first dimension explained 76.4% of the variance and that no other dimension explained more than 4.8%.

The person separation index of the BoHM was 4.21 meaning that the BoHM can separate infants into 6 different strata.

There was a significant difference in the EaHS of the lesser functioning hand and better functioning hand ($p < 0.001$). The correlation between age in months and the EaHS of the lesser functioning hand, the EaHS of the better functioning hand and the BoHM was weak ($r_s = 0.21$), strong ($r_s = 0.71$) and moderate ($r_s = 0.48$) respectively. There were no significant differences between the BoHM of different groups based on sex, affected side, country of scoring or country of origin.

Study I provides strong evidence for the internal scale validity of the results of the HAI for infants with asymmetric hand use. The HAI can be considered a unidimensional assessment tool and the main outcome of the HAI, the BoHM, can separate infants into six levels of ability which suggests that the BoHM may be responsive to change

6.3 NORMATIVE REFERENCE VALUES OF THE HAI (STUDY II)

Norm values were created by compiling 489 HAI-assessments of typically developing infants from Sweden and Italy. Descriptive statistics was used to create growth curves of the two sum scores, the EaHS and the BoHS. Both curves contained the mean and ± 1 and 2 SD for each month. The curves demonstrated the rapid development of object related hand use during the first 10 months of age. The Pearson correlation coefficient was used to investigate whether there was a correlation between the result in HAI and age. The correlation was strong between age and the EaHS ($r = 0.79$, $p < 0.001$) and the BoHS ($r = 0.85$, $p < 0.001$) showing that typically developing infants improve their results on HAI over age. The independent t-test indicated that there was no difference between the result of girls and boys on either the EaHS or the BoHS, as demonstrated by a p-value of 0.41 and 0.39 respectively. This indicate that there is not a need for sex specific age norms for the HAI. There was also no difference between the results of Italian and Swedish infants for either the EaHS ($p = 0.28$) or the BoHS ($p = 0.20$) indicating that there might not be a need for nationality dependent norm values. There was a statistically significant difference between the right and the left hand of 0.1 raw scores, however it was concluded that it would not be clinically relevant due to the small size.

For each item skill acquisition curves were created to visualize how the skill described in that item developed over the first 10 months of life (see Study II). For these curves data was dichotomized into Pass, if the infant scored a 2, and Fail, if the infant scored a 0 or a 1 and the curves demonstrated the percentage of infants passing the item for each month of age. The skill described in an item would be considered acquired if 90% of the typically developing infants

pass it. At the age of 10 months all skills except two, item 16 “Transfers objects in a sequence” and item 17 “Bimanual manipulation”, was acquired.

The normative reference values of HAI presented in Study II provides a reference to which we can compare the result of infants that we expect not to have a typical motor development. This provides us a tool that might be able to identify infants at risk of CP at an early age.

6.4 RELIABILITY OF THE HAI (STUDY III)

For the investigation of the test-retest and interrater reliability of the HAI 55 infants were assessed two times within a week. 30 of the infants were typically developing and 25 showed clinical signs of unilateral CP. The infants were recruited and filmed in both Sweden and Italy but all films were scored in Sweden by three experienced raters. To investigate the test-retest reliability the two films of each infant was scored in random order by the same rater. To investigate the interrater reliability, the first of the two assessments was scored individually by two of the scorers.

The infants with signs of unilateral CP was analyzed separately but all of the infants were also analyzed as a group. Intraclass Correlation Coefficient and CI was calculated for both groups on the BoHM and the EaHS, while ICC was calculated for each individual item. The Standard Error of Measurement (SEM) and Smallest Detectable Difference (SDD) were only calculated for the infants with signs of unilateral CP. For this calculation the EaHS of the better functioning hand and the lesser functioning hand were analyzed separately due to the different rates of development.

Test-retest reliability was excellent for the BoHM and the EaHS for both the full sample and the sample of infants at risk of unilateral CP, the same was true for inter rater reliability (see table 4).

Table 5. Reliability of the HAI

	Test-retest reliabilitet				Interrater reliabilitet			
	Full sample		AH		Full sample		AH	
	ICC	CI	ICC	CI	ICC	CI	ICC	CI
EaHS	0.99	0.99-1.0	0.99	0.98-0.99	0.99	0.98-0.99	0.99	0.98-0.99
BoHM	0.99	0.98-0.99	0.98	0.95-0.99	0.99	0.98-0.99	0.97	0.92-0.99

ICC Intraclass Correlation Coefficient, *CI* Confidence Interval, *AH* Infants with asymmetric hand use, *EaHS* Each Hand Sum score, *BoHM* Both Hands Measure

All individual items showed good or excellent reliability with an ICC >0.75 for both the test-retest and the interrater reliability, for both the full group and the group of infants with signs of

unilateral CP. For infants with clinical signs of unilateral CP the test retest reliability ICC was excellent for 82% of the items and all in all ranged from 0.81 to 0.97. For the interrater reliability ICC was excellent for most items (82%) and no item had an ICC that was lower than 0.81.

The SEM was 0.85 for the BoHM, 0.37 for the lesser functioning hand and 0.68 for the better functioning hand. The smallest change in score that is bigger than the measurement error and can be considered a true change, the SDD, was 2.36 HAI-units for the BoHM. This makes a change in 3 HAI-units, or 3% of the interval scale of 0-100 HAI-units, a real change. The SDD for the EaHS of the lesser functioning hand was 1.02 and for the better functioning hand it was 1.88 meaning that a change of 2 points raw scores (or 8% of the scale) for either hand can be considered true and not due to measurement error.

All in all, test-retest and interrater reliability is excellent for both the unimanual sum score, the EaHS, and the Both Hands Measure for both the full group and the group of infants with signs of unilateral CP. All individual item shows good or excellent reliability for both groups and a change of 3 HAI-units, for the BoHM, or 2 points raw score, for the EaHS, can be considered a true change.

6.5 SUMMARY OF RESULTS

The main results of this thesis are that HAI is the first assessment tool for infants 3-12 months of age that has an overall outcome measuring voluntary object related hand actions using both hands. HAI also measures each hand separately and gives a percentage of difference between the hands. There is strong evidence of the internal scale validity of the HAI for infants with an asymmetric hand use. The Rasch measurement model analysis demonstrated that the HAI measure one single construct. Furthermore, the HAI can separate infants into six different strata meaning that it can differentiate between infants of different levels of proficiency. The interrater and test-retest reliability of the HAI is excellent for the main outcome of the HAI, the BoHM, as well as for the sum score of the unimanual items, the EaHS. The reliability is good or excellent for all individual items for both the interrater and the test-retest conditions.

7 DISCUSSION

7.1 PSYCHOMETRIC PROPERTIES OF THE HAI

7.1.1 Aspects of validity

As has been emphasized earlier, a test cannot be considered valid per se, rather evidence of validity can be demonstrated for the result of the test performed on a specific target group (American Educational Research Association. et al., 1999). That is in this thesis we have not investigated the validity of the HAI but rather the validity of the results of HAI for infants with clinical signs of unilateral CP.

7.1.2 Evidence based on test content

The HAI was developed by a group of occupational therapists, physiotherapists and medical doctors with a large experience of children with cerebral palsy. The items and scoring criteria were constructed on base of knowledge of typical and atypical object related hand use in infants. Knowledge came from existing literature as well as systematic observations and clinical experience, and both items and scoring criteria were scrutinized and revised based on information collected when using the test-version of the HAI.

7.1.3 Evidence based on internal structure

The scores of an assessment is only useful if they reflect the underlying construct, that is if all the items measure the same underlying trait. This, the unidimensionality, of the HAI was investigated in Study I. The principal component analysis of the Rasch model measure analysis showed that the first dimension explained 76.4% of the variance of the result which is well above the limit of 60%. No other dimension should explain 5% or more and the second biggest dimension of the HAI explained 4.8% which is close to but still below what is recommended.

To collect more evidence of validity based on the internal structure of the HAI it would have been valuable to calculate DIF to investigate whether there was a difference between groups based on nationality. However, the samples of assessments from the different countries were very different in size (Swedish: 72, Italian: 32, Dutch: 20) and the sample was too small to perform analysis of DIF (Zwick, 2012). If future studies of the HAI include a Rasch measure model analysis with larger samples from different countries is recommended to investigate whether infants at the same ability level with different nationality has different response patterns for individual items.

7.1.4 Evidence based on response process

To investigate evidence based on response process of the HAI it is not relevant to interview the subjects to analyze their response patterns. However, it may be relevant to analyze what could influence how the scorers interpret the items and scores the infants. The excellent interrater reliability of the outcomes of the HAI indicates that there is no difference in how scorers interpret items. However, the scorers in Study III shared knowledge of aspects that could influence the judgement. For example, the scorers knew if the infant they were scoring was considered to be typically developing. Something else that could influence the scoring of an infant, that has been discussed in the research group, is if the therapist scoring the assessment also is involved in an intervention for the infant. In this case the therapist may have seen progress that is not captured by the HAI which could influence the intended interpretation of the scores.

7.1.5 Evidence based on consequences of testing

An expected consequence of assessing an infant using the HAI is that we can identify infants with clinical signs of unilateral CP. This can in turn lead to the infant receiving specific interventions and the parents receiving support something that is recommended in the literature (Novak et al., 2017). An assessment using the HAI also gives a quantitative result that can confirm something parents have already suspected (Baird et al., 2000). This information can be perceived as positive or negative.

7.1.6 Evidence based on relation to other variables

To investigate this aspect of the validity of the HAI, there would have to be another test that could be regarded as a gold standard measure to assess bimanual hand use in infants with signs of unilateral CP to compare it to. When the process of creating the HAI started this was not the case (Krumlinde-Sundholm et al., 2015), but since then the GRAB has been developed and published (Perez et al., 2016).

The GRAB is an assessment tool that holds many similarities to the HAI since they both share the same purpose; to detect an asymmetry in upper limb movements in the reaching and grasping of infants that are at risk of developing unilateral CP. However, while both the GRAB and the HAI are criterion referenced only the HAI is norm referenced (Krumlinde-Sundholm et al., 2017; Perez et al., 2016). This is important for two reasons, first of all knowledge of the degree of asymmetry that can be seen in typically developing infants is essential to correctly interpret the results of the HAI for an infant that is at risk of developing unilateral CP.

Furthermore, even if an infant at risk of unilateral CP demonstrates asymmetric hand use already at 3 months of age it is important to also scrutinize the use of the better functioning hand. It is not always clear at 3 months of age what type CP, if any, the child will later be diagnosed with, so we need to keep an open mind to the possibility that the infant may have a bilateral involvement. It is therefore of great value to also compare the better functioning hand to typically developing hand use.

The GRAB should only be used at set ages, that is 14, 16 and 18 weeks of age while the HAI can be used any time between 3 and 12 months of age (Krumlinde-Sundholm et al., 2017; Perez et al., 2016). This indicate that the HAI could be more feasible given the challenges in a busy neuropsychiatric clinic. For example, if an infant does not engage in handling the play objects during the HAI assessment the way that we would expect him/her to do for some unknown reason, maybe the infant is teething or getting sick, it is easy to re-schedule for a new assessment without having to take into account the infant's exact age.

It is also stated that the GRAB should be used in research only (Perez et al., 2016) while the HAI can be used both in research and in clinical work (Krumlinde-Sundholm et al., 2017). These differences between the GRAB and the HAI makes the HAI a more versatile assessment tool. Never the less it would be very interesting to compare the results of the HAI to the results of the GRAB to investigate the validity of the HAI in relation to other variables.

7.1.7 Reliability

When discussing reliability, it is important to keep in mind that there are no reliable assessments. What we do when we investigate reliability is rather that we investigate the reliability of the test scores of an assessment, that is how consistent the results are over repeated applications of measurement procedure (American Educational Research Association. et al., 1999). For infants with signs of unilateral CP the outcomes of the HAI showed excellent reliability for both test-retest reliability and inter-rater reliability as shown by the reliability coefficient. However, Koo et al (Koo & Li, 2016) emphasize that when you evaluate the reliability of a score you should not only concentrate on the reliability coefficient but rather the whole confidence interval. In other words, the results of an assessment cannot be said to have excellent reliability if all of the CI is not over 0.9. Even with that interpretation, both outcomes of the HAI show excellent reliability for both the Interrater and test-retest reliability with a lower limit of CI of 0.92 (table 4).

One important factor to take into consideration regarding the reliability of the results of the HAI is that all three assessors that participated in Study III had vast experience of the HAI. Two of the assessors had even taken part in the expert group that developed the HAI and participated in many discussions of the meaning of the items and scoring criteria of the HAI. It is therefore difficult to know to what extent the agreement in scores is accounted for by a common understanding of the items of the HAI that was not derived from the manual of the HAI. However, with the purpose to try to ensure a high degree of agreement between raters, a two-day rater training course is offered, which after approved results render a HAI rater certification. Future studies of reliability of the HAI should include more scorers and also some scorers that are less experienced.

7.2 NORMATIVE REFERENCE VALUES OF THE HAI

7.2.1 Cross-cultural use of the normative reference values

When the normative reference values in Study II were created we found no differences in the results of girls and boys or Swedish and Italian infants. Based on this it would be easy to conclude that the normative reference values of the HAI can be used in all of the western world. In clinical work it is not uncommon to uncritically use normative reference values for motor assessments of infants in other countries than they have been collected.

One assessment of motor skills for infants that is commonly used is the Alberta Infant Motor scales (AIMS). It is a gross motor assessment for infants 0-12 months that was developed in Canada in the 90s and is used worldwide (Piper, Pinnell, Darrah, Maguire, & Byrne, 1992). A study in 2006 compared the norm values of the AIMS, collected in Canada between 1990 and 1992, to the results of typically developing infants assessed in the Netherlands in 2004. The scores of the Dutch infants were significantly lower than the norms and 75% scored below the 50th percentile (Fleuren, Smit, Stijnen, & Hartman, 2007). It was hypothesized that this change may be due to new guide lines for infants to sleep in a supine position which in turn could cause delayed gross motor mile stones (Majnemer & Barr, 2005). However, a study comparing the original normative data to a new sample of typically developing Canadian infants in 2014 found no differences between the samples (Darrah, Bartlett, Maguire, Avison, & Lacaze-Masmonteil, 2014). The differences between countries regarding reference-values of the AIMS have been confirmed in several studies (Saccani & Valentini, 2013; Syrengelas et al., 2010) and it is hypothesized that the differences in results may be explained by differences between countries regarding economic and cultural factors (Saccani & Valentini, 2013).

These differences in gross motor skills in infants in different parts of the western world calls for afterthought before using the normative reference values of motor assessments in other countries than where the sample has been collected. The same factors that influence gross motor development may very well influence fine motor development as well. Economic factors that may influence motor development could for example be access to age appropriate toys to stimulate the infants bimanual hand use. A cultural factor that can differ between countries where you ordinarily place your infant, if there is a cultural habit of placing infants laying on a blanket on the floor or if it more common to keep infants sitting in a baby bouncer or if parents most often carry their infant. These differences in external trunk control in early infancy could perhaps influence the trajectory of both gross- and fine motor development. We could see a similar cultural difference between Sweden and Italy during the process of constructing the HAI, regarding at what age typically developing infants are expected to sit in a high chair versus a baby bouncer. Despite this there was no significant difference in the HAI results for the two countries neither for typically developing infants (Study II) nor for infants with asymmetric hand use (Study I). Nonetheless, the normative reference values of the HAI need to be used with consideration in other countries than Sweden and Italy and a need to establish country-specific normative reference values of the HAI should be investigated.

7.2.2 Skill acquisition curves in relation to existing knowledge

The skill acquisition curves were produced from the normative reference data of Study II. The focus of the study was to create the growth curves for the two sum scores, the EaHS and the BoHS. Quite early in the process it was discovered that many of the 10 months old infants achieved the maximum score for the BoHS. Data collection of infants 11- and 12 month olds therefore ended early since we wrongfully thought that the assessments would not provide additional interesting information. In later analysis we found that some of the skills, “Transfers objects in a sequence” and “Bimanual manipulation, were not yet acquired by all infants at 10 months of age. Given that result we now realize that it would have been interesting to have collected additional assessments of older infants as well to see the development of these skills until at least 12 month of age.

Below is a comparison between some of the skill acquisition curves to current knowledge of typical development of the fine motor skill that the item is intended to measure.

Voluntary grasp of objects exists from birth but becomes more manifest at four months of age (Charles, 2008; Gesell, 1934), which corresponds to the skill acquisition curve of item 5 Grasps from an easy position (figure 3).

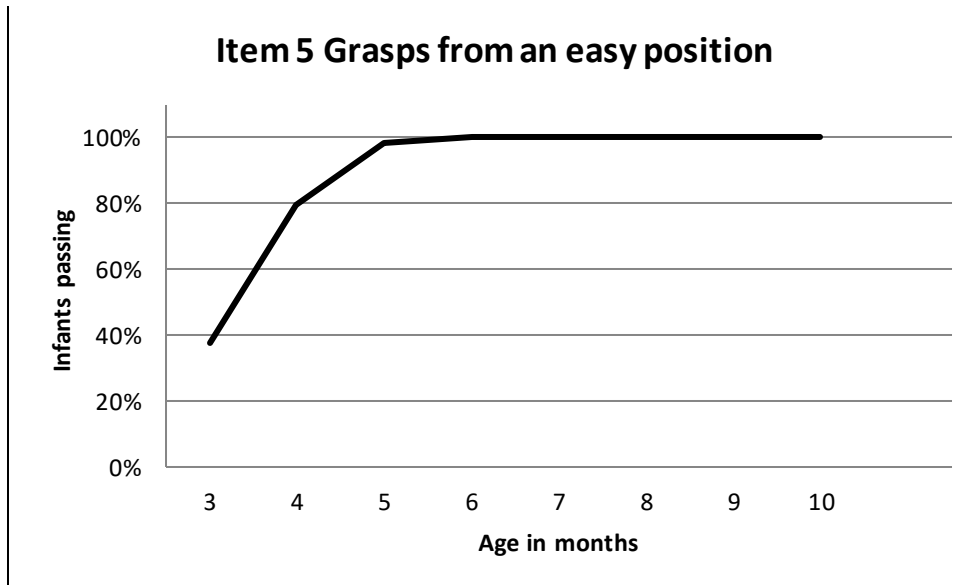


Figure 3. Skill acquisition curve of item 5 Grasps from an easy position.

Infants release objects purposefully at 6-7 months of age to move objects between the hands (Gesell et al., 1934; Pehoski, 2006; Pierce et al., 2009), this is also seen in the skill acquisition curve for item 15 Transfers between hands (figure 4).

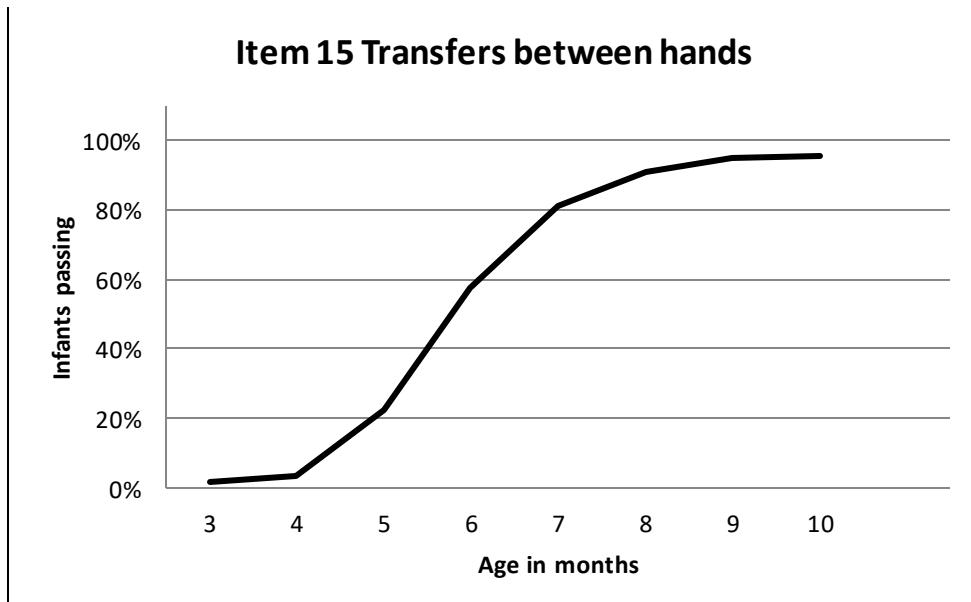


Figure 4. Skill acquisition curve of item 15 Transfers between hands.

Role differentiated hand use, when the hands do different things, starts to develop at 7 months of age but continues to develop throughout the first year of life (Charles, 2008; Kimmerle et al., 2010). In the HAI this is described in item 17 Bimanual manipulation and the skill acquisition curves corresponds to existing knowledge (figure 5)

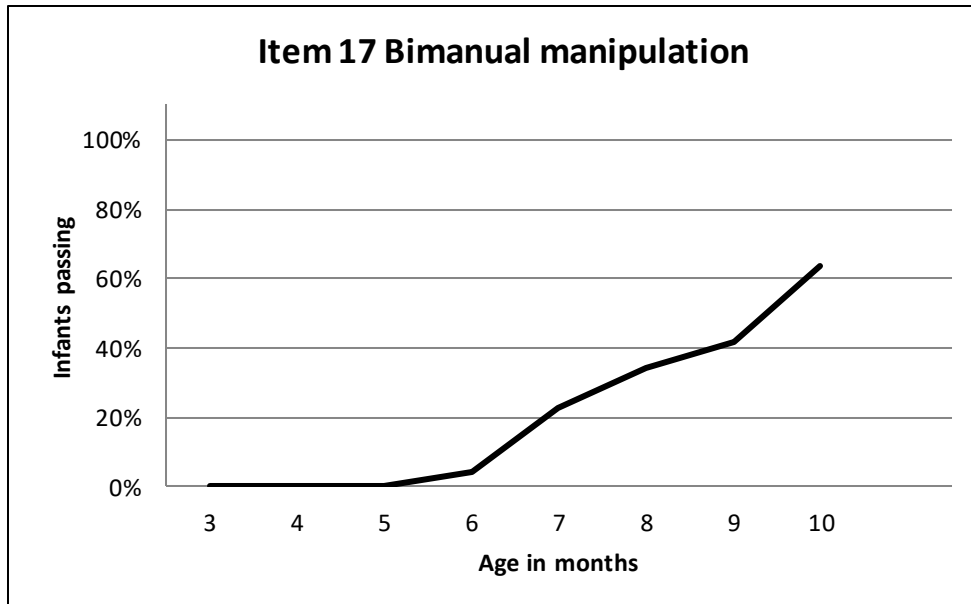


Figure 5. Skill acquisition curve of item 17 Bimanual manipulation.

One skill acquisition curve that appears to not correspond to the literature is that of item 7 Adjusts arm/hand position, which describes the infant's ability to pre-position the arm and hand to suite the shape and size of the object to be grasped (figure 6). Existing literature describes that infants start to adjust hand orientation to grasp objects at 5 months of age (Newell et al., 1993; von Hofsten & Ronnqvist, 1988) while the skill acquisition curve suggests that they do not (figure 6). However, when infants first start to adjust hand orientation they do it partially based on tactile feedback, that is after having touched the object (Witherington, 2005; von Hofsten & Ronnqvist, 1988) and only later they consistently demonstrate appropriate hand orientation based on visual feedback only (Witherington, 2005). In scoring this item with the HAI an infant that adjusts the arm and hand to grasp an item after having touched the toy would score a 1 for this item. Since data for the skill acquisition curves was dichotomized where only infants scoring a 2 would be considered as passing an item, therefore infants that adjust their grasp based on tactile response do not show on the skill acquisition curve.

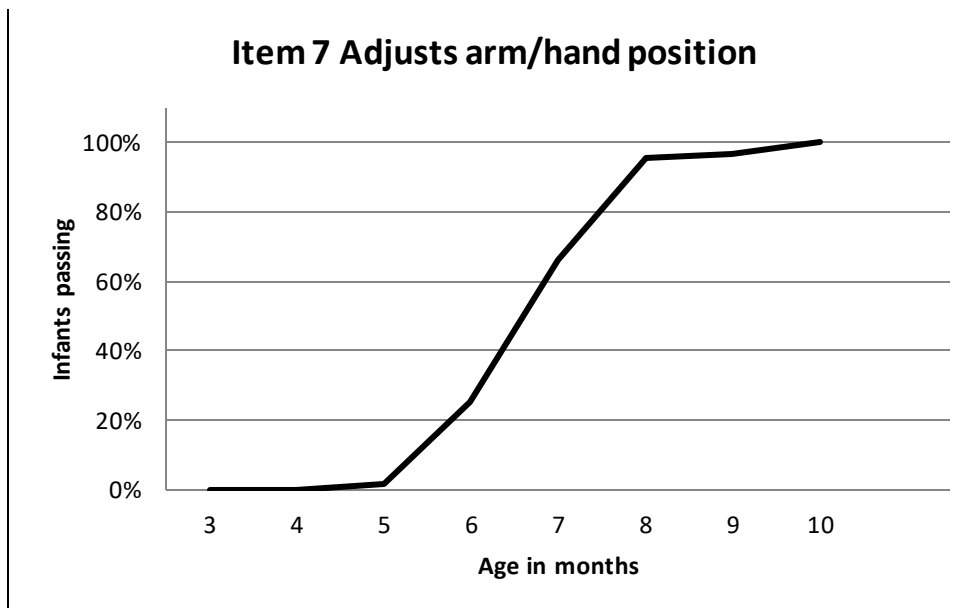


Figure 6. Skill acquisition curve of item 7 Adjust arm/hand position.

7.3 METHODOLOGICAL CONSIDERATIONS

7.3.1 Sample characteristics

The samples in all three studies exceeded the minimal required sample sizes (Chen et al., 2014; Hobart, Cano, Warner, & Thompson, 2012; Kranzler & Floyd, 2013; Norfolk et al., 2015). However, the samples of infants with signs of unilateral CP and infants with typical development were all convenience samples which means that the representativeness of the samples to the population is unclear. However, it is likely that fine motor skills of infants with signs of unilateral CP are more affected by the possible brain lesion than other factors like access to different toys. Furthermore, for the sample of typically developing infants an effort was made to achieve a spread in socioeconomic factors to improve representativeness. Even so a vast majority of the parents of the of typically developing infants were well educated.

7.4 FUTURE RESEARCH

This thesis has provided initial evidence of the psychometric properties of the HAI for infants with signs of unilateral CP as well as normative reference values. However, further studies of the measurement properties of the HAI are needed. The validity of the HAI in relation to other variables needs to be further investigated and the interrater reliability of the HAI should be further investigated in new studies including assessors with varied amounts of experience of the HAI. The cross-cultural use of the normative reference values of the HAI needs to be investigated to rule out a need for country specific age-norms.

An additional use for the HAI that could be investigated is if it is useful in relation to other groups of infants that has or is expected to have an affected fine motor development for example infants that are at risk of developing bilateral CP. Further developments of the HAI for use in infants with other types of unimanual disabilities like brachial plexus injuries or unilateral dysmelia would be an interesting development. Further studies to investigate the specificity and sensitivity of the HAI to predict unilateral CP would be of great value.

7.5 CLINICAL IMPLICATIONS

The clinical implications of this work should not be underestimated. The fact that HAI is the first assessment tool for infants that measures bilateral hand actions provides a new possibility to evaluate hand use and to examine longitudinal development. The current evidence of validity and reliability for infants with an asymmetric hand use provides an opportunity to, for the first time, measure and describe object related hand use for infants at risk of developing unilateral CP.

We now have a way to identify infants with signs of unilateral CP during the first year of life. Ongoing research shows excellent accuracy for prediction of unilateral CP for the HAI in combination with MRI and information of the gestational age and sex between 3.5 and 4.5 months of age (Ryll et al., 2019). This means that assessments with the HAI should be included in the follow-up programs of neonatal high-risk infants, hence providing an important role for the occupational therapists in the follow-up program. As of today occupational therapists are not even mentioned in the Swedish follow up program despite its emphasis on working in a multi-professional team and to identify infants at risk of CP as early as possible (Neonatalföreningen, 2015).

Given the stress of the importance of early interventions to improve motor skills in infants with unilateral CP, both nationally and internationally, this provides another field of work for occupational therapists, a field that is now uncharted, at least in Sweden.

8 CONCLUSIONS

As a result of the work included in this thesis there is now an assessment tool, the HAI, that can provide a measure of bimanual object related hand use in infants at risk of unilateral CP.

Researchers and clinicians now have access to an assessment tool that can quantify a possible asymmetry in hand use to identify infants at risk of unilateral CP.

Furthermore, there is now an assessment tool that can evaluate the effects of early fine motor interventions for infants at risk of unilateral CP.

This thesis provides evidence that the HAI is a valid measure of object related hand use in infants with signs of unilateral CP. The excellent reliability of the HAI shows that the HAI is a precise assessment tool that is sensitive to change.

9 SAMMANFATTNING PÅ SVENSKA

Redan vid 3 månaders ålder är det möjligt att se att barn som senare får diagnosen unilateral CP använder sina händer asymmetriskt i lek. Trots detta dröjer det ibland flera år innan barnet får sin diagnos. Detta gör att barnet får vänta på att bli inskrivet i Habiliteringen och få möjlighet att delta i interventioner för att förbättra den finmotoriska förmågan. Det behövs därför valida och reliabla bedömningsinstrument som tidigt kan särskilja barn med asymmetrisk handfunktion samt utvärdera effekten av tidig intervention.

Syftet med den här avhandlingen har varit att konstruera ett bedömningsinstrument som kan mäta bimanuell målorienterad handanvändning hos barn mellan 3 och 12 månader. Vidare har syftet varit att undersöka validitet och reliabilitet för det nya bedömningsinstrumentet för barn under 1 år med kliniska tecken på unilateral cerebral pares samt att skapa normativa referensvärden.

I avhandlingen beskrivs utvecklingen av, Hand Assessment for Infants (HAI), ett nytt bedömningsinstrument som mäter olika aspekter av målorienterad handanvändning. HAI innehåller 17 item varav tolv item är unimanuella, där höger och vänster hand poängsätts separat. Fem item är bimanuella där en sammanlagd poäng ges och alla item har tre skalsteg (0-2). Resultaten av de unimanuella itemen summeras till en Each Hand Sum score (EaHS) (0-24 poäng) för var hand för sig och ett asymmetriindex beräknas för att ge ett mått på asymmetrin i handanvändande. HAIs huvudsakliga utfallsmått är Both Hands Measure, en intervallskala på 0-100 HAI-units, som är ett sammantaget mått på hur barnet använder händerna bimanuellt i aktivitet.

Validiteten hos HAI för barn under ett år som uppvisade ett asymmetriskt handanvändande undersöktes genom Raschanalys. Resultatet visade att alla item i HAI mäter ett och samma underliggande konstrukt. Analysen visar också att HAI kan användas för att mäta barn med olika förmåga.

Normativa referensvärden skapades för HAI genom att sammanställa 489 HAI-bedömningar av typiskt utvecklande barn mellan 3 och 10 månader. Resultatet visade att typiskt utvecklade barn har en kliniskt signifikant men obetydlig skillnad mellan händerna. Normerna gör att det går att använda HAI för att identifiera barn under ett år med avvikande utveckling av handanvändning.

Reliabiliteten hos HAI undersöktes både mellan bedömare och för test-retest. Analysen visade att reliabiliteten hos HAI var utmärkt vilket demonstreras av höga reliabilitetskoefficienten (Intraclass Correlation Coefficient) som varierar mellan 0.97 och 0.99. Analysen av test-retest reliabilitet visar att den minsta skillnaden som kan mätas med HAI är 3 HAI-units.

Sammanfattningsvis visar studierna i den här avhandlingen att HAI kan användas för att göra giltiga och pålitliga bedömningar av hur barn under ett år använder händerna i aktivitet. HAI kan användas för att identifiera barn med asymmetrisk handfunktion och att utvärdera effekten av tidig intervention för barn som visar tidiga tecken på unilateral CP.

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